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CASE REPORT

Maxillary necrosis by Mucormycosis: A Case Report

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SUMMARY

A 50-year-old woman with type 2 diabetes was referred to the hospital with a 4-week history of a facial redness that proceeded to significant midface ulceration and bilateral vision loss. Her nasal bridge was completely collapsed, she had naso-palatal ulceration with black eschars on the mucosa, and her fasting blood sugar was significantly increased. Her blood sugar was controlled with soluble insulin through sliding scale, and surgical debridement was performed, which revealed fungal hyphae on histo-pathological assessment. Ketoconazole, an antifungal drug, was commenced immediately. She progressed slowly but steadily after that and her wound became clear with fresh granulation tissue.

Keywords: Mucormycosis, Uncontrolled diabetic mellitus

INTRODUCTION

Mucormycosis, also known as phycomycosis, is an uncommon invasive fungal infectious disease caused by Mucorales order and Mucoraceae family fungi. Following candidiasis and aspergillosis, it is the third most frequent angio-invasive & aggressive fungal infection[1]. It generally affects immunocompromised people and is only rarely observed in otherwise healthy people. Mucormycosis occurs in the immunocompromised host as a result of impaired immunity, resulting in fast expansion and infiltration of fungus in deep tissue structures[2].

There are multiple risk factors for mucormycosis that mainly consists of immunocompromised conditions of the body such as uncontrolled diabetes mellites, tumour conditions such as leukemias, chronic renal disease, organ transplant recipients, the patients on long-term corticosteroid and immunosuppressive medication, chronic liver disease and malnutrition[3]. Pathophysiological mechanism of mucormycosis involves inhalation of fungal spores through the nose or mouth, or even a skin incision. Patients with weakened cellular and humoral defense mechanisms may respond ineffectively to spores[4]. The fungus can directly extend to the paranasal sinuses and, as a result, to the orbit, meninges, and brain. Some mucormycosis individuals have no known risk factors. To avoid the significant morbidity and mortality linked with this illness, effective care of this lethal illness necessitates early detection of the disease and vigorous and quick medical and surgical therapies[5]. A case of maxillary mucormycosis in a diabetic patient is presented here.

CASE REPORT

A 50-year-old woman with uncontrolled diabetes mellitus was referred with a 4-week history of high-grade recurrent fever, severe throbbing headache, bilateral reddish nasal discharge, and progressive eye protrusion. The patient had a generalised facial edoema with pustules and blisters that impacted the right medial canthus of eye around a week following the onset of these symptoms. It expanded to her frontal and nasal area subsequently. Later, this developed into a large ulcer and mid-face necroses, with ill-smelling suppurative discharge from the lesions. The patient's vision in both eyes began to deteriorate quickly and she eventually lost her ability to perceive light. Prior to her presentation, she had experienced many bouts of brief loss of consciousness.

A general examination of the patient showed an ill-looking, emaciated, disoriented (GCS 11), and dehydrated female. The patient had drooping of eyelids, total drooping of her right upper eyelid with mucopurulent discharge, difficulty to open her eyelids, bilateral peri-orbital congestion, proptosis with no light perception bilaterally. The nasal cavity exhibited severe ulceration extending from both medial canthi, as well as a total collapse of the nasal cartilage. A 3.5x3cm round lesion in the anterior part of the hard palate was identified intraorally, revealing a blackish, necrotic mucopurulent material. There was also some crusting of the remaining nasal mucosa. Swelling of submandibular lymph nodes was also noted.

On investigations at admission, Packed cell volume was 25 percent, random blood glucose was 15mmol / L, white cell count was 25 x 109/L with 92 percent neutrophilia, platelet count was 514 x 109/ L, serum sodium was 1mmol/L, serum potassium was 4.5mmol/L, serum urea was 5.6mmol / L, and serum creatinine was 120umol / L.

The patient's computed tomography (CT) scan revealed soft tissue edoema in the right retroorbital and periorbital areas. The maxillary sinuses had significant soft tissue destruction causing obliteration, as well as bone damage of the medial side of the maxillary sinuses and the nasal septum. The ethmoidal air cells were also invaded by soft tissue, despite a normal chest x-ray.

The diabetes was quickly managed with soluble insulin through sliding scale, resulting in excellent blood glycemic control. In addition, the patient was given broad-spectrum antibiotics and intravenous hydration. After adequate blood grouping and cross matching, she received two pints of packed cells. The histopathological assessment was taken that demonstrated non-septate hyphae which is characteristic of mucormycosis.

Surgical debridement of necrotic material was performed thoroughly. As a result, she was treated by a team of otorhinolaryngologists, maxillofacial surgeons, ophthalmologists, and plastic surgeons. While Amphotericin B was being sought, she was commenced on ketoconazole. She made modest but steady recovery after starting antifungal and strict blood sugar monitoring, and her wound improved with robust granulation tissue formation.

DISCUSSION

Mucormycosis refers to a group of fungal diseases caused by Zygomycetes, which develop branching ribbon-like hyphae and reproduce sexually by forming zygospores. Pathogens can be found in abundance in fruits, soil, and excrement, as well as in the mouth cavity, respiratory system, and mouth of healthy people. Mucorales is a Zygomycetes subtype that causes this specific clinical illness. The fungi are normally non-pathogenic and only become pathogenic when the host's resistance is extremely low[6].

Mucormycosis infection is caused by asexual spores of the fungus. These small spores become airborne and settle on mucosal surfaces of an individual[7]. A phagocytic process will contain these spores in the most of immunocompetent individuals. If this process remains unsuccessful, germination and the formation of hyphae will occur. In immunocompromised patients, polymorphonuclear

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leukocytes are less successful in removing hyphae, and the infection gets settled[8].

Mucormycosis of the buccal cavity has two possible routes. The first is from disseminated infection, which is spread through inhalation, and the second is from direct wound contamination, which spreads to the viscera. When it comes through the nose or paranasal sinuses, the infection can progress to ulceration and necrosis of hard palate, with the afflicted area turning black in the majority of instances[8]. Clinical symptoms may emerge anywhere in the buccal cavity, involving the mandible, when the infection spreads from direct wound exposure. Cavernous sinus thrombosis, a major consequence of maxillary infections, is a notable distinction between infection involving the maxilla and infection affecting the mandible[9].

Diabetes mellitus alters the body's normal immune response to infection in a variety of ways. High glycemic index promotes fungal proliferation while lowering chemo taxis and phagocytic effectiveness, allowing otherwise harmless species to thrive in an acidic environment[7]. Mucormycosis caused by Rhizopus oryzae is more common in diabetic ketoacidosis patients because these organisms synthesize the enzyme keto-reductase that allows them to use ketone bodies[10]. The patient in this scenario was diagnosed with diabetes mellitus.

Oral, Rhino-cerebral, pulmonary, and cutaneous forms are the most common clinical presentations of mucormycosis, followed by enteric and miscellaneous types. In patients with unmanaged diabetes mellitus, the rhino-cerebral type of mucormycosis is most prevalent. Patients with rhino-cerebral type of infection have fatigue, headache, facial pain, edoema, and a low-grade recurrent fever. The infection usually starts in the oral or nasal mucosa, then spreads to the paranasal sinuses via the lacrimal and ethmoidal arteries. Mucormycosis can also potentially affect the retro-orbital region through direct extension[6]. Once fungal hyphae reaches the bloodstream, it can disseminate to vital viscera causing fatal infection.

Differential diagnosis of the lesions should include squamous cell carcinoma, basal cell carcinoma, chronic granulomatous diseases such as tuberculosis, and other severe fungal diseases.

On radiographs, opacification of the sinuses can be seen together with widespread effacement of the bone walls of the sinuses[5]. A CT scan with contrast or a magnetic resonance image scan can show bone degradation or destruction and help determine the severity of the condition[3]. The lesion has wide a-septate fungal hyphae that branch at right angles, according to histopathology. The same histopathological picture was demonstrated in this case.

Mucormycosis can be treated successfully of surgical debridement of the diseased region and systemic therapy with amphotericin B for three months if caught early. The management of the underlying disease is also critical in determining the treatment's success[6].

CONCLUSION

Mucormycosis is a virulent invasive fungal disease that can be caused by a variety of conditions, including uncontrolled diabetes mellitus, renal and liver failure, organ transplant, long-term use of corticosteroids, cancers such as leukemia, and AIDS. It can be induced even by simple dental procedures, such as tooth extraction, in a diabetic patient. More efforts should be made to diagnose this serious condition and treat the patient as soon as possible.

Declaration of patient consent: The authors attest to having gotten all necessary patient consent documents. The patient has given her agreement for her clinical information to be published in the journal by filling out the form. The patient is aware that her names and initials will not be published, and that while every effort will be taken to keep her identity hidden.

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